

SARCOMA OF THE TONGUE.

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IN his monograph on 'Diseases of the Tongue,' which was published in 1885, Mr. Butlin remarks that "primary sarcoma of the tongue must be regarded as an exceedingly rare form of tumour." So little material was he able to collect, that he felt it quite useless to attempt to write an account of this disease; but he adds, "Now that attention has been drawn to its extreme rarity, it is possible that some instances which have been observed, and not yet recorded, may find their way into surgical literature." In this paper I propose to record a case which occurred last year in Guy's Hospital under the care of Mr. Howse, to whom I am indebted for permission to publish the notes, and to append abstracts of such cases as are reported in the various medical journals. An examination of the surgical reports of Guy's Hospital for the last twenty-five years has supplied but three cases, including that which forms the subject of this paper.

CASE 1 ('Guy's Surgical Reports,' 1888).—Joseph P—, æt. 65, admitted under Mr. Howse for a swelling in the mouth, which he first noticed a year ago. It was then the size of a horse-bean, and was situated about the middle of the tongue; but it has gradually enlarged ever since, though without causing any pain. He says he has lost weight lately. Family history and previous history very good.

On admission there is in the substance of the tongue a

globular tumour, harder than the tissues of the tongue, and not fluctuating. It is most apparent on the left side of the frænum. This globular mass is separated by a groove from another swelling which extends along the left side of the floor of the mouth from the frænum to the last molar tooth, is softer than the former, and semi-fluctuating. These two swellings appear to be connected deeply in the submaxillary region, for with the forefinger on the floor of the mouth and the thumb under the ramus of the jaw they seem to move as one. In the submaxillary region, too, is an enlarged, freely movable lymphatic gland.

The patient can swallow easily and speak clearly, but the movements of the tongue are somewhat impeded by the tumour. There is no pain. The surface is not ulcerated.

Antisypilitic treatment was adopted for three weeks, but as there was no visible improvement an operation was recommended. Under an anæsthetic the left cheek was incised from the angle of the mouth backwards for an inch and a half, the tongue was drawn forwards, and was divided with scissors along the median line for the anterior two thirds of its length, and the left portion removed, including the growth and the mass in the floor of the mouth. There was free hæmorrhage requiring many ligatures; afterwards the wound was painted over with iodoform in ether, and the incision of the cheek was closed.

The subsequent treatment consisted in painting over the raw surface in the mouth with iodoform every hour for the first day, and less frequently afterwards; and the patient was fed with milk through a tube passed over the back of the tongue. Within three weeks of the operation he was discharged quite well.

Fifteen months after the operation the patient was again examined, and a large mass found in the left submaxillary region, which had been slowly increasing in size. There was also a swelling below the zygoma on the right side of the face, which appeared to be a secondary deposit. In the floor of the mouth and the remainder of the tongue there was no recurrence. He complained of much discomfort in the chest, and had had a severe attack of bronchitis, but there were no physical signs of secondary disease in the lungs.

Examination of the tumour.—The parts removed by operation were found to contain two masses of new growth, one the size of a walnut occupying the site of the sublingual gland, and the other embedded in the substance of the tongue. A vertical longitudinal section of the tongue showed that the latter mass was spherical in shape, an inch and a half in diameter, and very soft in structure. It appeared moderately well defined, the muscular tissue being displaced by it; its upper limit was half an inch below the dorsal surface of the tongue; in front it nearly reached the frænum linguæ, while its outer surface was immediately beneath the mucous membrane of the floor of the mouth. There was no ulceration of the mucous membrane in any part. What has been described above as the sublingual portion was very closely connected with the mass in the tongue, and was probably an extension of the disease. Its relation to the sublingual mucous gland could not be determined by dissection or histological examination; hence it was suggested that the latter had either not been removed at the operation, or had already been destroyed by the growth.

Microscopic sections were prepared from different portions of the tumour, after the tissues had been embedded in celloidin. They were composed of small round cells, remarkably uniform in size, and held together by a little granular material. There was no true stroma, but the sections were permeated with thin-walled vessels which in some degree supported the cells. A few strands of muscular and fibrous tissue ran through the sections, and served to divide them up into large areas of cells, but were not sufficiently abundant to render the tumour firm. Within these strands one or two nerve-bundles were seen in transverse or oblique section. Further, among the cells themselves tiny pieces of muscular fibres were occasionally found, and the method of invasion by lines of cells between the muscle-fibres was easily made out. The sections were carefully searched for parasites, but without success. It was, therefore, concluded that the growth was a true round-celled sarcoma, which had begun probably in the connective tissue of the substance of the tongue, and gradually destroyed and replaced its muscular tissue; in other words, that it was a primary sarcoma of muscle affect-

ing the tongue, and resembling in its structure and development such new growths in other regions of the body.

CASE 2 ('Guy's Surgical Reports,' 1867).—Albert W—, æt. 2, was admitted under Mr. Durham for a swelling on the tongue which his mother had first noticed seven weeks before. It had much increased in size of late, but there was no pain in it. The child snored loudly in its sleep. The family history was very good.

On admission.—There was a tumour on the dorsum of the tongue, to the left of the median line and about one inch and a half behind the tip. It was whitish, of a soft consistency, and measured one inch in diameter. There was no enlargement of the glands in the submaxillary region. Deglutition did not appear to be affected, and there was no pain.

The child remained under observation, and two months after admission, as the tumour had increased in size and was interfering with swallowing, a portion of the tongue, including the tumour, was removed with an *écraseur*, though it was afterwards found that not quite the whole of the tumour had been taken away. A fortnight after the operation recurrence *in situ* had been so rapid that the tumour was even larger than before. It was somewhat nodulated and very soft, and extended backwards as far as the finger could reach, feeling as if attached to the left anterior pillar of the fauces. It also encroached upon the floor of the mouth, and could be felt externally as a mass deeply situated in the left submaxillary region.

The patient was removed from the hospital, and died shortly afterwards. No autopsy was obtained. Microscopical examination of the tumour showed that "it was composed of delicate fine fibre-tissue, with cells exhibiting all the transitions in shape from round cells, through oval and spindle-shaped, to simple fibres."

CASE 3 ('Guy's Surgical Reports,' 1873).—Charles J—, æt. 10 months, a healthy child with good family history. The mother first noticed a sore under the tongue at the age of seven months, which gradually developed into a "lump," and bled frequently. When admitted into the hospital under

Mr. Howse there was a white button-shaped eminence on the under surface of the tongue, extending from the floor of the mouth to the tip in the median line. It had a flattened surface, raised rounded edges, and a spongy appearance. After removal sarcomatous tissue was stated to exist in the base of the tumour.

The further history of this case is not known, as the patient did not return to the hospital.

CASE 4 (Jacobi, 'Amer. Journ. of Obstetrics,' vol. ii, 1870, p. 81).—Thomas K—, æt. 11 weeks. He was born of healthy parents, and his brother (aged three years) showed no signs of syphilis. A few hours after birth the mother noticed a swelling of the tongue, but it did not interfere with suckling. It slowly enlarged, however, and was punctured, though nothing but blood was evacuated.

When the child first came under observation there was a tumour of the tongue the size of a walnut, occupying the left half of the tongue from the median line to the margin, and half an inch from the tip. It was well rounded inferiorly, encroaching much on the floor of the mouth, but the upper surface was less regular. It was moderately firm and elastic, of a bright red colour, and its surface was marked with a distinct network of large capillaries. The lymphatic glands were not enlarged, and in all other respects the child was healthy, though deglutition was now impeded owing to the increase in size of the swelling.

The tumour and the adjacent portions of the tongue were removed with a galvanic écraseur, and no hæmorrhage resulted. A fortnight later the wound was healing rapidly.

In the report of the microscopical examination the cells are described as mostly spindle-shaped, with oviform, oblong nuclei, and very uniform in size and shape; hence the tumour was considered to be a spindle-celled sarcoma. The external (? *peripheral*) portions of the tumour contained much muscular and interstitial tissue and capillaries, but less in proportion of the constituents of the growth proper.

No further history of this case has been published.

CASE 5 (Bleything, 'New York Med. Journ.,' 1888, xlvii, p. 683).—A lad, æt. 17, had a painful "ulcer" of the tongue

on the right margin half an inch from the tip. It was covered with a small mass of granulations, and there was some induration of its base. The patient attributed the sore to a broken tooth. As repeated applications of the cauterly failed to cure, and as the swelling enlarged and its base became harder, the affected portion of the tongue was removed; six years afterwards the patient was in excellent health, with no sign of recurrence.

The microscopical examination of the diseased tissue showed "areas of grouped cells, with new areas of infection around gorged vessels;" and after due consideration, and a not unnatural hesitancy between granulation and sarcomatous tissue, the tumour was declared to be a sarcoma of the tongue.

CASE 6 (Godlee, 'Trans. Path. Soc.,' vol. xxxviii, p. 346, 1887).—A woman, æt. 24, came under observation with a swelling on the under surface of the left half of the tongue near the tip. It had only been noticed for five weeks. The tumour was hard, the size of a cobnut, and formed a slight projection upon the dorsum of the tongue; in the centre of the swelling there was a grey slough as large as a shilling. It was removed with scissors, and the wound healed rapidly. On inquiry it was ascertained that there was no recurrence of the disease three years after the operation.

Histologically the growth was an adeno-sarcoma. A large proportion of the tumour was made up of sarcomatous tissue, with round and spindle-shaped cells and many vessels. The glandular elements consisted of acini, varying much in size and shape. In the centre of the tumour was a small salivary calculus.

It was suggested that the tumour had probably arisen in connection with the mucous gland described by Blandin and Nuhn,

CASE 7 (Heath, 'Trans. Path. Soc.,' vol. xx, p. 157, 1869).—The patient was a man, æt. 60. The disease had existed six months. On examination "there was a ragged ulcerated surface in the floor of the mouth between the tongue and the lower jaw on the left side. This extended to the side of the tongue, and with the finger a large mass could be felt in the

substance of the tongue, reaching beyond the median line, and to about two inches from the lips." There was constant pain in the tongue, but the patient's health was otherwise good. The lymphatic glands were not affected.

The anterior half of the tongue was removed freely. Twelve years afterwards the patient was in good health, and the stump of the tongue was sound.

The specimen on section presented the appearances of a medullary cancer. The report of the Morbid Growths Committee is as follows: "The tumour was for the most part embedded in the under and outer part of the left side of the tongue, not far from the tip, but a portion of it was prolonged downwards among the muscular attachments of the tongue, and not improbably involved a part of one of the salivary glands. The tumour was small, well circumscribed, had an opaque white colour, and on scraping yielded not so much a juice as abundant minute fragments of apparently solid tissue. The part of it embedded in the substance of the tongue was spongy; the lower part was homogeneous and solid. On microscopical examination, the cell-elements of which the tissue was composed were in some places more or less regularly round or oval; in others they were elongated, or presented one or more tail-like processes." The remainder of this lengthy report is too vague for the purposes of exact histology, but it may be noted that there is no mention of the presence of bird-nest formations, of epithelial ingrowths, or of a definite alveolar structure. A drawing which accompanies the report most nearly represents a small spindle-celled sarcoma. The committee expressed their opinion that "the tumour was essentially a cell-growth invading and displacing the normal tissues, the cell-growth forming equally the more obvious cell-structure of the tumour and its fibrous portion."

CASE 8 (Albert, 'Wiener medizinische Presse,' 1885, p. 171).—The patient, a woman *æt.* 56, stated that she had suffered from difficulty in swallowing for nearly a year. A month after the onset of this symptom she noticed a swelling on the tongue, which slowly but steadily enlarged. On examination a smooth roundish tumour was found to be attached to the

root of the tongue, and it extended upwards almost to the hard palate, concealing the isthmus faucium. The attachment of the tumour involved the whole breadth of the tongue, to which it was firmly fixed, and its outline was well defined. Its consistency was moderately hard throughout, except in one or two spots which felt decidedly soft. Excision of the whole tongue was performed through the submaxillary region. Death took place on the eighth day after the operation from bronchitis and lobular pneumonia. The microscopical examination of the tumour showed that it was a round-celled sarcoma.

CASE 9 (Poncet, '*Lyon Médical*,' 1888, lviii, p. 95).—The report of this case states that the patient was a man, æt. 32, who came under observation for a tumour of the tongue which had been growing for eight years. The lower jaw was divided and the tongue excised. The tumour weighed 400 grammes, and was considered to be a fasciculated sarcoma originating in the substance of the tongue, very probably in the submucous and intermuscular cellular tissues.

In reply to an inquiry respecting the further history of this case, Prof. Poncet very kindly informed me that his patient was in excellent health fifteen months after the operation, with no sign of recurrence of the tumour. The publication of a more detailed account of this case is contemplated.

CASE 10 (Hutchinson, '*Med.-Chir. Trans.*,' vol. lxxviii, p. 311).—The patient was a man, æt. 22. A swelling was first noticed on the left side of the tongue at the age of ten years. It continued to increase without causing inconvenience, except from its size, until he was twenty-one. The tumour consisted of a large rounded mass, deeply embedded in the tongue, and wedged in between the sides of the lower jaw. The tip of the tongue was free, and could be moved on the surface of the tumour, but with this exception the whole of the organ was involved. The mucous membrane of the posterior two thirds of the tongue on the left side was covered with a coarse papillary growth, continuous with the substance of the tumour beneath, and not ulcerated. A year later the growth had increased considerably, and the inconvenience

was unbearable. It was removed by division of the lower jaw and the application of an *écraseur* round the base of the tongue just in front of the epiglottis. A rapid recovery followed, and the patient continued well for many months; subsequently, however, the growth recurred in the floor of the mouth, and spread rapidly, causing death, partly by pressure and partly by hæmorrhage, two years and a half after the operation. There was no evidence of glandular enlargement. On examination the tumour was almost globular in shape, measuring two inches and a half in diameter, and not encapsuled. It weighed seven ounces. It completely replaced the substance of the tongue, save a small portion three fourths of an inch in length at its tip. The posterior part of the new growth showed a greyish fibrous structure, which was divided into loculi by white bands; in front it was softer and more vascular. The muscular tissue of the tongue was inseparably attached to the tumour. Histologically the growth was a round-celled or lympho-sarcoma, consisting of large rounded alveoli filled with lymphoid cells of uniform size, enclosed by bands of fibrous tissue and intersected by finer strands of the latter. Between the cells a delicate reticular tissue was seen in parts. Here and there in the substance of the tumour muscular fibres, atrophied by pressure, were observed.

CASE 11 (Butlin, '*Lancet*,' 1887, vol. i, p. 623).—A man about 40 years of age was admitted with a swelling of the left half of the tongue. Two months previously he had noticed a soreness of the tongue in the region described, but the swelling had only appeared a month before admission, and during that interval it had increased rapidly.

When first examined a smooth, soft, elastic swelling was found in the substance of the left half of the tongue. It was about the size of a racket-ball, and extended from the junction of the anterior and middle thirds to the root of the tongue, bulging on its upper and under surfaces, but not crossing the median line. There was no ulceration of the mucous membrane over it. The tongue could not be protruded from the mouth, but the patient complained of no pain except on eating. His general health was good. A hard enlarged gland was detected in the submaxillary region.

Large doses of iodide of potassium were administered, but as no diminution in the size of the tumour took place the left half of the tongue was excised three weeks after admission. The gland under the jaw was not removed. Five weeks later the patient was quite well, and could talk plainly. There was no glandular enlargement and no sign of recurrence.

At the present time, more than three years after the operation, the patient is in excellent health, and the condition of the mouth is very satisfactory.

On examination of the parts removed a soft white circumscribed tumour was found, with the appearance of a capsule at one spot. It was oval in shape, measuring one inch and a half in length by an inch in breadth, and was situated immediately beneath the mucous membrane of the middle third of the tongue on the left side. Microscopically it consisted of sarcoma-cells, which were large, round, and granular. The matrix was generally homogeneous, but in parts granular. Numerous small blood-vessels permeated the section. The tumour belonged to the class of lympho-sarcomata.

In addition to the above cases two examples of connective-tissue tumours of the tongue have been recorded by Eve ('Trans. Path. Soc.,' vol. xxxvii, p. 223), but as they have no clinical history, and differ widely in anatomical details from the tumours at present under consideration, no further mention of them need be made. Butlin ('Diseases of Tongue,' p. 259) refers to one case of tumour of the tongue, sections of which he had the opportunity of examining, but he did not see the tumour itself, and knew nothing of the clinical features of the case. "It presented the characters of a round-celled or lympho-sarcoma," he adds.

Barker ('Holmes' System of Surgery,' vol. ii, p. 576) mentions a case in which a somewhat pendulous growth upon the dorsum of the tongue was removed and soon recurred. Several growths of the same kind appeared subsequently on the skin of different parts of the body. He regarded the case as in fact an example of multiple sarcomata, the dorsum of the tongue participating in a general disease of the cutaneous surface of the body. Finally, a case is recorded by Santesson, of Stockholm, of an angeo-sarcoma of the tongue, but I can

give no further details, as the original description is inaccessible.

Having thus dealt with the literature of the subject as fully as possible, it only remains for me to review shortly the cases quoted above, and, in conclusion, to make a few remarks on their clinical and pathological bearings. Of the eleven cases described in more or less detail, Nos. 1, 2, 7, 8, 9, 10, and 11 have some points in common, and will be considered together. Case 5, though declared to be a sarcoma, was probably inflammatory, as it began in an irritation-ulcer; it was composed of "areas of grouped cells, with new areas of infection around gorged vessels;" and there were no signs of recurrence at the end of six years. Case 6 must be considered as an adeno-sarcoma of a mucous gland, rather than a primary tumour of the substance proper of the tongue, and should therefore be placed in a different category from that of the majority of the cases here recorded. Of Case 3 the evidence is insufficient to enable one to form an opinion as to its nature; it might have been an inflamed nævoid growth. Lastly, Case 4, though quoted by all writers on this subject, is too vague for classification, and, as the accuracy of the diagnosis has been doubted by such a competent authority as Mr. Butlin, it may be put on one side.

Of the remaining seven cases it may be said that the disease was of the substance of the tongue and not of the surface, though a prominent mass was eventually developed in several instances; that the mucous membrane covering the tumour was not ulcerated (with one exception), and therefore pain was absent; that the lymphatic glands were not affected in the cases in which this point is mentioned, or only in the later stages, as in Case 1, where the submaxillary swelling was probably in the main an extension of the growth; and that local recurrence after operation was rapid in only one out of the six cases (excluding No. 8), while in three others it had not taken place at the time of publication. These remarks will be made clearer by the following scheme :

Case.	Age.	Length of history.	Situation, &c.	Results.
I	65	1 year	In substance of left half towards root; one gland enlarged	Recurrence within one year.
II	2	4 months	Middle third towards dorsum; glands normal	Rapid local recurrence. Death.
VII	60	$\frac{1}{2}$ year	In substance towards root; glands normal	No recurrence at end of 12 years.
VIII	56	1 year	Whole breadth of root of tongue	Death a week after operation.
IX	32	8 years	In substance	No recurrence at end of 15 months.
X	22	12 years	Deeply embedded, involving whole organ; glands normal	Death from local recurrence $2\frac{1}{2}$ years after operation.
XI	40	7 weeks	In substance, left half; glands normal	No recurrence at end of 3 years.

The cases are too few to enable any useful conclusions to be drawn from the age and sex of the patients; the duration of the disease also varies within wide limits. But with regard to the latter point it is worth while to note that Case 9, which existed eight years, was a fasciculated sarcoma, and therefore probably of much slower growth than the remaining cases; and in Case 10, of still longer duration, Mr. Hutchinson was of opinion that the disease had begun in a congenital mole or nævus, and only developed "morbid tendencies" at a much later date. The similarity in histological structure of the tumours is not a little remarkable, for with one, or perhaps two, exceptions they were of the round-celled or lympho-sarcomatous type. Of the two exceptions, one was a fasciculated or spindle-celled sarcoma, and the other (Case 2) was of the mixed-cell variety. This point is of importance in the question of diagnosis, and will be referred to subsequently.

In a recent publication¹ on malignant diseases of the tongue and lips, Esmarch states that in his experience "the majority of tumours which are commonly recognised as sarcoma and extirpated belong to the syphilomata, and can

¹ Abstracted in 'Centralblatt für Chirurgie,' 1889, No. 29.

be cured by antisyphilitic treatment, whilst without this they frequently pursue a malignant course." The conclusions he draws are that the sarcomata of muscle-tissue are mostly syphilitic in nature (syphilomata); that as the tongue consists chiefly of muscular tissue, so syphilomata appear in it frequently, and that in many cases the origin of tumours (particularly sarcoma) is dependent upon a predisposition inherited from syphilitic ancestors. This theory derives little support from the cases here recorded, partly from the imperfection of the clinical accounts, and partly from the fact that in two of the cases (1 and 11) antisyphilitic treatment failed. Apart from the minute structure of the tumours, the fact of their recurring locally, and sometimes rapidly, is, I venture to think, opposed to such a view. Deep gummata of the tongue, with which sarcomata are most likely to be confounded, are said to be often multiple and of long duration, and will probably be associated with other syphilitic lesions, scars, ulcers, enlarged glands, &c. Admitting, then, the necessity of caution in the diagnosis of tumours of the substance of the tongue, I cannot at the same time doubt that malignant growths, which would be recognised by the majority of pathologists as sarcomata, do actually occur in the tongue, and stand in need of operative treatment. But whether they are due to ancestral syphilis, or represent a malignant transformation of an old syphilitic lesion, there are not enough facts at present to decide.

The difficulties of diagnosis, however, are not confined to syphilis. Chronic abscess and actinomycosis, and some innocent tumours too, will present swellings which must be distinguished from sarcomata. With regard to actinomycosis there are at least four cases of primary disease on record.

Ullmann¹ relates two such, and Hochenegg² and Hacker³ one each. These cases presented nodules in the substance of the tongue, near its tip or borders, which were at first hard, but softened as they enlarged, and caused no pain. In one instance the tumour was the size of a pigeon's egg. The mucous membrane was not affected, but the submaxillary

¹ 'Wiener medizinische Presse,' 1888, No. 50.

² *Ibid.*, 1887, No. 16.

³ 'Wiener medizinische Wochenschrift,' 1885, No. 17.

lymphatic glands were enlarged. The source of infection could not be determined in all. The diagnosis appears to have been settled by puncture and microscopical examination of the contents. Though actinomycosis of the tongue has long been known in the lower animals, its occurrence as a primary disease in the human tongue is not yet generally recognised.

The treatment of sarcoma of the tongue scarcely calls for comment. Slow recurrence after free removal by operation is not peculiar to this region. But the preponderance of the round-cell type of growth in the examples quoted suggests the importance of an early and free excision.

In conclusion, I submit that there is a true bill for sarcoma of the tongue, but that it is a very rare disease as compared with epithelioma of that part, there being only four specimens preserved in the various London museums. The difficulties in its diagnosis are considerable, so that an adequate anti-syphilitic treatment should first be adopted in all cases. The resemblance which primary actinomycosis bears to it should not be forgotten, though operative treatment is required for both. In structure it is most frequently a round-celled growth, beginning in the substance of the tongue, the cause of which has yet to be determined.

Note.—Dr. Waller, of Rugby, writes to say that at the present time (August, 1890), nearly two years after the operation, the patient, Joseph P—, is enjoying fair health. There are large masses of secondary growth on both sides of the neck, also below the right zygoma and in the left sub-maxillary region, but there appears to be no recurrence in the mouth.